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The experience of type 2 diabetes self-management in adults with intellectual disabilities and their caregivers: A review of the literature using meta-aggregative synthesis and an appraisal of rigour

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Background People with intellectual disabilities (ID) experience significant barriers to diabetes self-management (DSM), yet there remains a paucity of research within this population. An overview of the literature on people with ID and their caregivers' experiences of living with and self-managing type 2 diabetes (T2D) is provided.

Methods Meta-aggregative methods were adopted to synthesise results, and an appraisal was reported of rigour.

Result A total of eight studies met the inclusion criteria and four themes were extracted: i) "Frustration over lifestyle adjustments"; ii) "Limited understanding and inadequate educational resources"; iii) "Limited training and knowledge in staff"; iv) "Potential for effective DSM with appropriate support".

Conclusion Current support is inadequate to meet the needs of people with ID and their caregivers self-managing diabetes. Structured education to improve health literacy and diabetes knowledge in people with ID is required, together with training for caregivers which leads to a culture of nurturing autonomy.

Background

Type 2 Diabetes (T2D) is a global epidemic associated with poor glycaemic control. There are complex and severe health risks associated with the disease, including renal failure, blindness, amputation due to circulatory problems, and cardiovascular risks such as stroke and myocardial infarction. There are therefore severe cost implications to health services from diabetes. Diabetes UK reported a cost of £23 billion in 2010/2011, of which 8.8 billion was for T2D (Hex et al., 2012). This figure is projected to almost double by 2035.

The prevalence of T2D in people with intellectual disabilities (ID) compared to the mainstream population is unclear. The most recent review of diabetes prevalence rates in

people with ID was carried out by MacRae et al. (2015), who identified 22 studies. This provided a mean prevalence rate of 8.3%. This is compared to a global prevalence rate of 8.5% in the mainstream population (World Health Organisation, 2014). However, prevalence rates vary widely across studies, ranging from 0.4% to 25%, reflecting several reporting issues, such as failure to report differences in type of diabetes and a reliance on self-reports from people with ID, their family members or caregivers. Therefore, evidence suggests that prevalence rates may be higher for people with ID than for the general population.

The lack of demographic information, such as living arrangements (absent in 15/24 of the studies in MacRae et al., 2015), is notable, as this may have elucidated for example whether people with ID who live in isolated situations are less likely to receive diabetes screening, hence not being screened for diabetes and receiving a diagnosis and treatment. This suggests that there is a lack of consistency over reporting details in studies about people with ID and diabetes.

An additional focus of the MacRae et al. (2015) review, and an earlier review by McVilly et al. (2014) was on DSM in people with ID. In the mainstream population, DSM is seen as the most important and cost-effective approach towards T2D care (Davies et al., 2008). However, as these two reviews highlight, there are several issues for people with ID self-managing T2D with regard to making lifestyle changes after diagnosis. A limited number of studies presented findings on the experiences of DSM in people with ID. Subsequent to these two reviews, several recent studies have offered further insight into the self-management experiences of people with ID and their caregivers, such as facilitated self-management and the discussion of findings in relation to theoretical models. The findings from these, as well as from less recent studies, are important as the views and experiences of people with ID and their caregivers have the potential to inform and guide the development of intervention programs aimed at assisting this population. Such interventions are urgently

required, given the estimated global prevalence of T2D, inequalities, and additional support needs of many people with ID (Cooper et al., 2015).

There is therefore a further need to provide a cohesive overview on how people with ID are managing their diabetes, in terms of adapting their lifestyles and adherence to diabetes management plans, and the level of support that is required to ensure effective compliance. The primary purpose of this review is to identify and appraise the scope and extent of the qualitative research studies which focus specifically on the DSM experiences and perceptions of people with ID and their caregivers, and to present the findings as themes using a narrative synthesis approach. As a secondary aim, an appraisal of the rigour of these studies is provided. The two systematic reviews of studies in this area (McVilly et al., 2014, and MacRae et al., 2015) have provided a quality appraisal using the Effective Public Health Practice Project (Thomas, Ciliska, Dobbins and Micucci, 2004). This tool has been designed to evaluate quantitative criteria found in trials, such as sampling representativeness and drop-out rates. As such, it is not appropriate for evaluating qualitative studies, which explore the complexities of experiences and subjective meaning of phenomena. Therefore, a qualitative appraisal tool (Elliot, Fischer, and Rennie, 1999) has been used to evaluate rigour.

Aims

- 1) To provide an overview of the common themes in qualitative studies in relation to the self-management of T2D in adults with ID.
- 2) To provide an appraisal of the methodological quality and theoretical application of the findings from the studies to describe the overall rigour of the literature.

Methods

Literature search strategy

Elements of the Joanna Briggs Institute (JBI) approach of meta-aggregation were used to inform the methods of this review, following the guidelines provided by Lockwood, Munn, and Porritt (2015). This approach is phenomenological and pragmatic, enabling an in-depth synthesis of complex phenomena in a small body of literature, which retains the authors' original interpretation of the findings in the synthesis.

The following inclusion criteria were applied: studies with a specific focus on people with ID self-managing type 2 diabetes; published in peer-reviewed journals; written and published in English. Due to the paucity of the literature, searches were designed to be as broadly inclusive as possible no restrictions were applied to the dates of studies or to further demographic restrictions such as age, gender and level of disability were not made.

The following studies were excluded: studies which focused on specific learning difficulties, such as dyslexia and dyspraxia; studies which did not focus on self-management of type 2 diabetes; studies not published in peer reviews; studies not published in English

Search terms and data sources

A review of the published literature focusing on self-management of T2D was carried out systematically using the key-words, “Intellectual Disabilities” OR “Learning Disabilities” OR “Mental Retardation” AND Type 2 Diabetes OR Diabetes Mellitus AND Self-management OR Self-care in the following databases: PUBMED; CINAHL; MEDLINE; PsychINFO; Psychology and Behavioral Sciences Collection. Further hand searches were carried out of study references.

Literature appraisal strategy and tools

The studies in this review were evaluated using the qualitative methodological guidelines by Elliot, Fischer, and Rennie (1999). These guidelines serve as a system for ensuring robust evaluation of qualitative studies by providing a set of flexible, evolving criteria. The 7 guidelines include; 1) Owning one's perspective: The author explicitly states their own values, interests and assumptions; 2) Situating the sample: The author provides a detailed background description of the participants; 3) Grounding in examples: Examples of the data are provided, such as transcript excerpts; 4) Providing credibility checks: The author verifies their coding and analysis; 5) Coherence: The data is integrated into a coherent narrative; 6) Accomplishing general vs. specific research tasks: The authors state the limitations of the data in terms of generalizability when using limited samples or singles case studies; 7) Resonating with the readers: The manuscript is presented in a way that enables the reader to judge it as an accurate description of the phenomena when all guidelines are taken into account. The studies were initially screened by the main author, then across the research team for consistency and accuracy.

Data extraction

Elements of the JBI meta-aggregation approach were adopted for data extraction. This involved a multi-phase process in which general data was first extracted, including study origins, participant demographics and methods (Lockwood et al., 2015). The details of this

phase are presented in Table 1, supplementary materials. Three of the studies were drawn from the same sample of people with ID (Hale et al., 2011; Trip et al., 2015; Whitehead et al., 2016). Trip et al. (2015) and Whitehead et al., (2016) supplemented this sample with support staff, which was also a shared sample. Therefore, to prevent overlap of reporting of demographic details, shared characteristics of these samples were not synthesised. However, as all three of these studies had a different focus and analysis yielding different results, each one was used in the later stages of synthesis described below.

Data synthesis

Following extraction, synthesis of the main findings of the studies was carried out. This involved extracting every finding from each study and accompanying this with an “illustration”, which included either a participant quotation in the form of a transcript excerpt, or an observation of the researcher supported by the number of participants. Ratings were then made based on the congruence of the finding and the “illustration” as i) “Unequivocal”; ii) “Equivocal”; iii) “Unsupported” (Lockwood et al., 2015). This stage of analysis was verified by the second author, and differences between ratings were resolved through discussion. A summary of the findings and ratings are provided in Table 2, supplementary materials.

The final stage of the synthesis involved organising the “Unequivocal” and “Equivocal” rated findings into categories. “Unsupported” findings were not included in the

synthesis. Categories were then synthesised into themes. These themes were discussed and agreed with the second author. A summary of this process is presented in Table 3, supplementary materials.

Results

A total of eight papers were identified, as illustrated in the flow diagram (*figure 1*) below.

Two of these were conducted in The Netherlands: Cardol, Rijken, and van Schrojenstein et al. (2012a); Cardol, Rijken, and van Schrojenstein Lantman-de Valk, (2012b); two in the UK: Dysch, Chung, and Fox (2012); Rouse and Finlay, (2016); three in New Zealand: Hale, Trip, and Whitehead (2011); Trip, Conder and Hale (2015); Whitehead, Trip, and Hale (2016); one in Australia: Rey-Conde, Lennox, and McPhee (2005).

Insert Figure 1 here:

Sample sizes and participant characteristics

Study characteristics are presented in *Table 1*, supplementary materials. The sample sizes ranged from $N = 4$ (Dysch et al., 2012) to $N = 67$ (Rey-Conde et al., 2005). The range of sample sizes reflected the different aims and methodologies, with smaller samples in exploratory studies which recruited people with ID only (such as Dysch et al., 2012 and Hale

et al., 2012), and larger studies which recruited people with ID and their supporters in order to evaluate theoretical constructs (Trip et al., 2015). Although the sample size of $N = 4$ in Dysch et al. (2012) was small compared to the other study sample sizes, IPA studies typically range from $N = 1$ to $N = 15$ (Petkiewicz and Smith, 2012), therefore this was not necessarily an indicator of poor quality. However, recruitment of people with ID can present challenges, such as the ethical requirement to use third parties when approaching vulnerable adults, as highlighted by Hale et al., (2011). This issue led to the common approach across the studies of using a convenience sample, which may have detracted from the representativeness.

A total of three studies recruited people with ID only (Cardol et al., 2012a; Dysch et al., 2012; Hale et al., 2011). There were two studies which recruited caregivers only (Cardol et al., 2012b; Trip et al., 2015). These focused on the experiences of support staff and keyworkers in supporting people with ID to self-manage T2D. Three studies recruited people with ID and support staff (Rey-Conde et al., 2005; Rouse and Finlay, 2016; Whitehead et al., 2016). Rey-Conde et al., and Whitehead et al., explored the experiences and perceptions of T2D self-management. This latter study used the data from Hale et al., (2011) and Trip et al., 2015). Rouse and Finlay explored the concept of responsibility between people with ID and their caregivers.

A total of three studies reported the level of ID (Hale et al., 2011; Dysch et al., 2012; Cardol et al., 2012a). The levels described were “Mild” (mean 7.33, SD 3.5), “Moderate” (mean 5, SD 2.82), and “Unknown” ($N = 7$: Cardol et al., 2012 only). Rouse and Finlay reported “mild or moderate” without specifying which of these levels applied to their seven participants, yet detailing that two of these participants had autism and none had specific conditions such as Down’s Syndrome. The lack of inclusion of moderate and profound participants may reflect communication difficulties in this population which can lead to difficulties in giving consent and being able to give detailed responses. ID levels are based

upon cognitive assessment of IQ, however it is unclear in these studies whether these groupings were based on formal cognitive assessment.

The living status of people with ID was reported in three studies (Hale et al., 2011; Dysch et al., 2012; Cardol et al., 2012a). The reported living statuses were “residential care” (mean 5, SD 5.2), “supported independent” living (mean 3.3, SD 3.2), and “independent living” (mean 1, SD 0). The high number of participants in residential care, together with the high number of mild ID reported, suggests that across the studies most participants were receiving a high level of support, despite having low support needs. The data from these studies therefore may not be representative of the majority of people with ID self-managing diabetes, as the challenges for people with higher support needs who live in more isolated situations was not explored.

A total of three studies reported the number of participants with Type 1 diabetes (T1D) or T2D (Hale et al., 2011; Dysch et al., 2012; Rouse and Finlay, 2016). There were slightly fewer participants with T1D (mean 3.3, SD 2.3) than with T2D (mean 5, SD 3.6). Although self-management was described in all of the studies, there were only two which specifically reported the self-management approaches of individual participants within the methods section (Cardol et al., 2012a, Whitehead et al., 2016). These included “diet alone” (mean 2, SD 1.4), “medication” (mean 7, SD 1.4), and “insulin” (mean 6.5, SD 2.1). Wider reporting of this information could have led to a greater understanding of the prevalence and efficacy of these approaches in people with ID.

The time since diagnosis was reported in 3 studies (Hale et al., 2011; Dysch et al., 2012; Cardol et al., 2012a). As there were differences in how these durations were presented, it is not possible to provide descriptive comparisons. However, in each study there were a significant number of participants with a long-term diagnosis than those who had been diagnosed recently. This included over 5 years since diagnosis, (Cardol et al., 2012a) over 10

years since diagnosis (Hale et al., 2011), and mean 17 years (Dysch et al., 2012). This trend reflects the inclusion of type diabetes participants, for whom diagnosis would be more likely to be detected at an early age, as well as suggesting more progressed stages of T2D, where self-management would be typically dependent on medication or insulin. The small number of participants who were managing their diabetes through diet alone may reflect that there is a higher prevalence of T1D and advanced stage T2D, where medication and insulin control is a necessity. However, this may also reflect the struggle of the participants to adapt their lifestyle to diabetes; this is reflected in the themes described below.

Themes highlighted through synthesis

Following the review and analysis of the eight studies, the following four themes were identified: i) "Frustration over lifestyle adjustments"; ii) "Limited understanding and inadequate educational resources"; iii) "Limited training and knowledge in staff"; iv) "Potential for effective DSM with appropriate support".

1. Frustration over lifestyle adjustments

People with ID expressed negative feelings with regard to living with and self-managing diabetes, such as fear, insecurity, and being overwhelmed (Rey-Conde et al., 2005). These were accompanied by a sense of frustration over changes to lifestyles, in particular dietary restrictions (Hale et al., 2011) medication regime adherence, and carer dependency (Dysch et al., 2012).

Participants also made statements which Cardol et al. (2012a) described as a sense of loss in relation to food choices. These feelings were occasionally accentuated by shared

living situations in which social comparisons to others without diabetes were made, an issue also present in "Potential for effective DSM with appropriate support". It is important that supporters of people with ID are aware of such feelings, as these may impact on mental wellbeing and long term adherence to DSM behaviours.

The social setting of the participants, rather than diabetes itself, appeared to contribute to frustration at times, and act as a barrier to DSM. Cardol et al (2012a) provided an example of a participant with ID who no longer engaged in physical activity due to lack of transport provided by their organisation. A participant in Dysch et al. (2012) described experiencing social stigma around injecting insulin at work, which they were concerned would be seen drug-use. This may have impacted on their sense of restricted lifestyle. However, it was reported by Dysch et al. (2012) that social comparisons could also facilitate understanding of DSM. People with ID's understanding of diabetes is discussed in the following theme.

2. Limited understanding and inadequate educational resources

This theme synthesises categories in which the authors described limitations of diabetes DSM in people with ID, as described by their statements and the perceptions of caregivers.

Dependence upon caregivers for DSM was either described directly by people with ID (Rey-Conde et al., 2005, Dysch et al., 2012 and Hale et al., 2011) or inferred by the authors' in their interpretation of people with ID's statements (Dysch et al., 2012). In addition, caregivers made statements which described limited DSM competence in the people they supported (Cardol et al., 2012b, Hale et al, 2011, and Rouse and Finlay, 2016). As is highlighted further below in "Attitudes, management of dilemmas and impact of social setting", these statements may be indicative of care attitudes as well as DSM competencies.

The studies indicated that overall people with ID had a limited understanding of diabetes, which sometimes impacted on DSM skills. Cardol et al., (2012a) reported that the

consequences of having diabetes were only perceived as serious when insulin injections were required, although it is important to acknowledge that this study included participants with Type 1 diabetes. Insulin injections are necessary regardless of progression in Type 1 diabetes, whereas it can be possible to control type 2 diabetes with diet, exercise and medication. Therefore, there was a lack of clarity through not clarifying this distinction in relation to the participants' statements. However, Cardol et al.'s (2012a) conclusion that there was a relationship between understanding and DSM is a key-finding reflected throughout the themes, highlighting the need for diabetes education for people with ID.

Other findings indicated that where educational resources were available, they were not appropriate for people with ID due to being in formats that were not accessible (Hale et al., 2011). Rey-Conde et al. (2005) also reported that information on diet was insufficient leading to confusion. It is therefore important that educational resources are appropriately tailored for people with ID.

3. Limited training and knowledge in staff

Following on from the lack of knowledge of people with ID highlighted above, a lack of training and knowledge in staff was also described across the studies. Rey-Conde et al. (2005) reported that staff and families found limited staff training to be a barrier to effective self-management support. Rey-Conde et al. (2005) attributed some of these difficulties to high levels of staff turnover, exacerbated by limited training and experience. However, information was not provided about the level of training and qualifications of staff, or their duration of employment, making it difficult to conclude this finding.

Cardol et al. (2012b) reported that there were varying levels of knowledge according to whether participants were nurses, who had more specialist diabetes training, or care workers. Participants who were social workers reported that diabetes training often needed to

be of their own volition, and that management occasionally impeded training. Care workers often did not perceive the seriousness of diabetes and, similarly to the people they supported, diabetes care was not a prominent part of care unless administering insulin was required (Cardol et al., 2012b).

A limited awareness of available resources also appeared to reflect poor staff training; Hale et al. (2011) reported that some carers were unaware of diabetes management plans for an individual they supported. Cardol et al., 2012b and Trip et al., (2015) reported an overall inconsistency in staff knowledge and training, and to some this was frustrating as they could not rely upon the competence of their colleagues (Trip et al., 2015). This inconsistency extended to training in supporting others to be autonomous in DSM (Cardol et al., 2012b).

Caregivers' descriptions of their role may be reflective of the barriers and challenges they face in supporting DSM in people with ID, but may also describe attitudes which highlight a need for further training. Trip et al. (2015) reported caregivers who described themselves as "lifestyle police", who took a prohibitive approach in order to reduce the risk of unhealthy diets. Concern about this risk was also described by caregivers in Rey-Conde et al. (2005). People with ID were described as "lazy" and "passive" by caregivers in Cardol et al. (2012b), possibly reflecting a dispositional attribution. Whilst these caregivers may have been in an expert position to evaluate the characteristics of the people they supported, there were approaches reflected in the final theme, which describe viable and empowering alternatives to the prohibitive approach. It is notable however, that Rouse and Finlay (2016) reported people with ID describing themselves as "lazy". This was interpreted as a defensive repertoire against incompetence and dependency. In this case, caregiver training may also be required to support and enhance positive self-perceptions in people with ID.

Caregiver dilemmas also reflected a potential need for further training in caregivers. These dilemmas were between enabling autonomy and reducing risk, and were described in

four studies (Cardol et al., 2012b; Rouse and Finlay, 2016; Trip et al., 2015; Whitehead et al., 2016). As with the attitudes described above, a difference in caregiver approach was highlighted in how these dilemmas were resolved, and an absence of flexible, creative solutions may reflect a need for further training.

4. Potential for effective DSM with appropriate support

Although feelings of frustration were described by people with ID in the first theme, there were also descriptions of positive feelings which highlighted the potential for successful DSM. Cardol et al. (2012a) interpreted their findings to suggest that Self-efficacy in people with ID was linked to DSM. Self-efficacy is the confidence in oneself to achieve tasks and overcome barriers (Bandura, 1977). This confidence in DSM was also reported in Rey-Conde et al. (2005) and Rouse and Finlay (2016).

Dysch et al., (2012) interpreted that participants showed an understanding of diabetes through the language they used and through their awareness of physical symptoms, comorbidities and fluctuating states. This suggests a more sophisticated level of understanding than as was suggested above in “Limited understanding and inadequate educational resources”. In addition, Hale et al. (2011) described participants who were not only aware of the benefits of exercise to DSM, but also suggested a buddy-system as a potential exercise facilitator. People with ID may therefore potentially have the insight for understanding and subsequently self-managing diabetes, which could be enhanced through education.

Some studies highlighted the recognition of caregivers that whilst there were often limits to people with ID’s competence in DSM, there were also areas of strength. Caregivers

in Rey-Conde et al. (2005) reported that despite there being a need for concern positive exercise and dietary and choices were made by people with ID. Similarly, Hale et al. (2011) reported caregivers who, despite the need for support, described the awareness of blood sugar change symptoms in the people they supported, and also their awareness of recording times. Structured education was suggested as a solution to DSM barriers such as cognitive impairment, suggesting that change was perceived as possible.

Caregivers also showed a potential for effective DSM was through the flexible and creative support of caregivers. Whilst it was highlighted above in “Limited training and knowledge in staff” that there was a dilemma of protection versus enabling autonomy, some caregivers provided solutions to this dilemma. Whitehead et al. (2016) described a process of “negotiated autonomy”, in which levels of support were adjusted in relation to the present needs of the people they supported. Participants were described as almost fully autonomous until situations such as medication adjustments or changes in living arrangements arose, presenting a need for higher support. However, as these situations passed, autonomy was restored where possible. Conversely, Cardol et al. (2012b) described a reduced restriction of dietary choices and provision of healthy alternatives on special occasions such as birthday parties. Although this may reflect a prohibitive approach overall than in Whitehead et al. (2016), such a flexible approach may reduce the frustrations of people with ID which were expressed above.

Finally, Rey Conde et al. (2005) described aspects of support which could facilitate independent self-management. These included providing prompts and supporting planning, which may potentially facilitate overcoming barriers and reducing relapses. Such strategies suggest that there are caregivers who look beyond controlling approaches and that this is sometimes reflected in practice. The importance of opportunities for shared knowledge of how to nurture independence was highlighted in Whitehead et al. (2015), although time was

acknowledged as a barrier to this. However, investing in such approaches could potentially lead to a more consistent application of flexible and creative solutions.

Study Quality Evaluation

Owning perspective

The majority of the studies did not include information on the researcher background, with the exception of Rey-Conde et al. (2005), who described the research team as being part of a clinical service for adults with ID, and that expectations were low due to observing poor self-management. However, it was difficult to conclude whether this suggested a possibility of confirmation bias, as other aspects of evidence, such as transcript excerpts, were less well described.

Situating the sample

The majority of the studies provided detailed demographics, with the exception of Rey-Conde et al. (2005). However, situational information about individual participants was only provided in one study (Dysch et al., 2012). Further personal, details such as recent life events or family circumstances may have enabled the reader to evaluate the accuracy of the themes, although studies may have been restricted from seeking and providing information due to the need for protecting participant anonymity.

Grounding in example

All studies included ample relevant transcript excerpts, as reflected by the number findings in the meta-aggregative synthesis. In general, the transcript excerpts of people with ID were more substantial than those of the caregivers. This is counterintuitive, given the communication difficulties of people with ID; richer examples could potentially have been provided from the caregivers.

Credibility checks

The majority of the studies described credibility checks to some degree, with the exception of Rouse and Finlay (2016). Rey-Conde et al. (2005) described triangulation, through checking transcripts against audio recordings. However, there was no mention of additional auditing, for example using a second researcher. Similarly, Trip et al. (2015) described multiple readings of transcripts, but did not triangulate or verify through another researcher. Hale et al. (2011), Dysch et al. (2012), Cardol et al (2012a&b) and Whitehead et al. (2016) described a review process where coding and themes were discussed until consensus was reached. The general adherence to this guideline across the studies gives credibility to the analyses.

Coherence

A total of four studies conformed to this guideline (Dysch et al., 2012; Cardol et al., 2012a&b; Trip et al., 2015). Rey-Conde et al (2005) and Whitehead et al. (2016) did not provide descriptions of the hierarchical structure or supportive figures. Hale et al. (2011) described sub-themes but did not support these with figures. These variations may reflect differences in journal stipulations, such as figure inclusions.

Accomplishing general versus specific research tasks

Rey-Conde et al. (2005) did not acknowledge the limitations of their sample sizes and nature. Cardol et al. (2012b) and Trip et al. (2015) did not fully report the limitations of their sample in terms of generalizability, however both studies reflected that their findings were consistent with other studies in the literature.

Resonating with the reader

The most commonly missed criterion was “Owning perspective”, with only one study including this (Rey-Conde et al., 2005). Other studies read less well due to sparse or brief transcript excerpts, unclearly described analytical procedures, or lack of description of thematic structure. However, these qualities did not impact highly on the overall resonance of the studies, as experienced by the reviewers.

Discussion

The primary aim of this review was to provide an overview of studies which qualitatively represent the experiences of people with ID and their caregivers living with and self-managing T2D. The first theme “Frustration over lifestyle adjustments”, described findings which highlighted challenges, and barriers for people with ID. These provided support for the findings of McVilly et al. (2014) and MacRae et al. (2015). With regard to sense of loss, the present review has offered a further insight by looking at this theme alongside the participants' duration since diagnosis. This highlighted that adjustment difficulties can be long term for people with ID and that continued support may be necessary. The social setting of the participants was also highlighted as impacting upon mental wellbeing, as positive and

negative social comparisons were made. As highlighted in the demographics, the majority of the participants lived together in residential settings, where they were likely to live alongside each other because of having a learning disability, rather than having diabetes. It has been reported that people with ID sometimes make downward social comparisons with each other as a self-esteem protecting mechanism (Paterson, McKenzie and Lindsay, 2012). This may further impact on frustrations regarding social comparisons to peers who do not have diabetes, and supporters should be aware of the potential for this. The suggestion in Hale et al. (2011) of a “buddy-system” for exercise, although positive, may also be subject to the complexities of social comparisons and social stigma.

The second theme, “Limited understanding and inadequate educational resources” highlighted gaps in the diabetes knowledge in people with intellectual disability. In terms of people with ID's lack of education, the fact that people with a long history of diabetes continue to struggle to understand the nature and implications of the disease suggests that there is a need for ongoing education, as well as at diagnosis or even prior to development. With this in mind, preventative approaches aimed at younger adults and adolescents with ID (such as Yates et al., 2012) could provide an early foundation which establishes good health behaviours and reduces the risk of developing diabetes. Given the lower health status of people with ID compared to the general population it is likely that, regardless of glycaemic levels, they may have multiple risk factors for diabetes, such as obesity, sedentary lifestyles and poor diets, regardless of glycaemic levels. Therefore it would be appropriate for this group to receive structured education aimed at reducing diabetes risk factors (such as Biddle et al., 2015 and Yates et al., 2012), though such programs may need to be appropriately tailored.

Although structured education programs are commonplace in the mainstream population, none are presently available for people with ID. Walwyn et al. (2015) have

reported a trial protocol which describes the development a nurse-led DSM intervention for people with ID. Taggart et al. (2017, in press) reported the success of a feasibility study adapting the DESMOND structured education program for people with ID, although logistical challenges were such as transportations were reported. Indeed, in the mainstream population long-term adherence to such interventions has been described as in decline for similar reasons (Coates et al., 2017). The present review indicates that the consistency of caregiver support is integral to DSM, and this is likely to also impact upon sustained behavioural change.

Quality and consistency of caregiver support was described in “Limited training and knowledge in staff”. As the majority of the participants in the studies highlighted in this review were in residential care, the level of support was high. People with ID who do not live in supported settings are less likely to access staff with general medical training such as nurses, however they may require a high level of training due to diabetes-related medication administration needs. It is therefore possible that the current literature does not fully represent the lack of training in the care of people with ID with diabetes, and further research is required on how to support people with ID who have diabetes in other settings. It was highlighted that further training and resources are required for caregivers, and this should be incorporated into structured educational programs. A study by O’Leary, Taggart and Cousins, (2016) on organisational barriers to health promotion in people with ID found that there was a lack of cultural ethos within residential settings. This finding may account for the inconsistency of approaches highlighted within the review, and creative approaches which foster autonomy may unfortunately be limited to individuals, rather than being present at an organisational level. It is therefore appropriate to suggest that DSM education is extended to managers and senior staff, so that such approaches are more widely implemented.

The final theme, "Potential for effective DSM with appropriate support" highlighted the strengths of people with ID and their caregivers in DSM, thus providing potential facilitators. Areas in which people with ID felt confident were identified, and it was suggested that Self-efficacy (Bandura, 1977) may be an applicable theoretical construct. As with the findings above, these facilitators may be limited to people with a mild ID in a residential setting. However, the fostering of creative and flexible approaches may potentially enable caregivers to support autonomy in people with higher support needs across a range of settings. It is therefore important that structured education provides training for caregivers which steers away from prohibitive approaches.

The secondary aim of this review was to provide a qualitative appraisal of the selected studies. The overall quality was not high, as indicated by the Elliot et al. (1999) criteria who emphasise the importance of acknowledging the researcher position. This was not commonly adopted across the studies, and may reflect general lack of adherence to reflexivity in methodological approaches such as Interpretative Phenomenological Analysis and Grounded Theory. However, the relationship of the researcher to the participants is a commonly applied criterion in appraisal tools, (for example CASP, 2013), and is important to include so that assumptive positions are clear.

Findings were rigorous in terms of the provision of extracts and validity checks. This is reflected in the number of findings included in the present review's analysis (see *Table 2*, supplementary materials). However, there was an absence of description of analytical procedures. The Elliot et al. (1999) tool acknowledges that qualitative research is necessarily pragmatic, presenting a need for flexibility, and it is therefore acceptable for methods to be unique and innovative. However, this leads to a greater need for transparency, so that techniques can be evaluated and repeated.

Strengths and limitations

This review has taken a rigorous approach, and the result is a high-quality synthesis and evaluation of the narrow pool of studies on people with ID living with and self-managing diabetes. The number of included studies is a limitation, as is the inclusion of studies which also included Type 1 diabetes which led to difficulties in isolating findings applicable to type 2 diabetes. However, adopting meta-aggregative methods enabled an in-depth analysis which is suited to a small number of studies (Lockwood et al., 2015).

Conclusion

These findings, although mainly restricted to people with mild ID living in residential care, highlight that people with ID are currently not sufficiently supported to self-manage type 2 diabetes. To address this, tailored education needs to be developed and delivered to improve diabetes knowledge and health literacy, but also to build upon areas in which people with ID are confident and develop Self-efficacy. Training is required to ensure the consistency of creative and flexible approaches from caregivers, which suggests that structured education should be attended by caregivers alongside the people they support. Furthermore, managers and senior staff should be provided with training to foster autonomy at an organizational level. Further research is required into the nurturing of autonomy for people with higher support needs and beyond residential settings.

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Figure 1: Flow chart of study selections

